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Esophagogastric fistula complicating Nissen fundoplication

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ABSTRACT

Esophagogastric fistula or double-lumen esophagus is a rare condition. There have been fewer than 15 reported cases in adults and only one reported case in the pediatric population. Esophagogastric fistulas typically develop in patients with preexisting gastrointestinal reflux, esophagogastric surgery, esophageal ulcers, or carcinoma. Our case involves a 5-year old girl presenting with odynophagia and nocturnal cough who had a prior Nissen fundoplication. She was found to have an esophagogastric fistula. Conservative management with esophageal dilatation and proton pump inhibitors was not successful. However, a repeat Nissen fundoplication with fistula repair relieved the patient of her symptoms.
Level of evidence: Level V; Case report.

Esophagogastric fistula is a very rare anatomic finding. We report a case of esophagogastric fistula in a 5-year old girl who was evaluated for odynophagia and nocturnal cough after Nissen fundoplication at the age of 2.

1. Case report

1.1. Past medical and surgical history

A 5-year old girl born at 26 weeks gestation with past medical history significant for grade 1 intraventricular hemorrhage, persistent ductus arteriosus, and prolonged neonatal intensive care unit stay presented with odynophagia and nocturnal cough after Nissen fundoplication at age 2.

At the age of one, she was investigated for oral aversion and failure to thrive. At that time, an upper gastrointestinal contrast study showed no abnormalities; however, endoscopy indicated distal esophagitis. A gastrostomy was placed to supplement her diet.

At 2 years of age, she had severe regurgitation. A repeat contrast study confirmed gastroesophageal reflux. She underwent Nissen fundoplication with a 360° fundal wrap over a 28 Maloney bougie. No crural repair was necessary. The fundoplication was secured with four interrupted 4-0 silk sutures. A superficial “bite” from the anterior aspect of the esophagus was also included in each “bite.”

Three days postoperatively, the patient reported dysphagia and sialorrhea. Nine days later, endoscopy revealed obstruction of the distal esophagus at the site of the fundoplication. No specific treatment was administered, although gastrostomy tube feeds were continued, her symptoms had spontaneously improved. A postoperative contrast study showed a tight fundoplication with an esophagogastric fistula in the form of an inverted “Y” configuration of the distal esophagus. However, she did well clinically and the gastrostomy was removed six months later.

At the age of 4 she had adenotonsillectomy, for sleep disordered breathing complicated by adenotonsillar hypertrophy.

1.2. Chief complaint

She now presents at age five complaining of “pain in her throat” after eating, central chest pain, nocturnal cough, and morning hoarseness. Contrast esophagogram (Fig. 1) demonstrates a Y-shaped deformity of the distal esophagus. Flexible endoscopy identified two outlets at the gastroesophageal junction. One outlet is narrow and could not accommodate the gastroscope. The esophagogastric fistula was also visualized with retroflexion of gastroscope at the gastric fundus. The esophagus proximal to the gastroesophageal junction had significant erosive esophagitis. A

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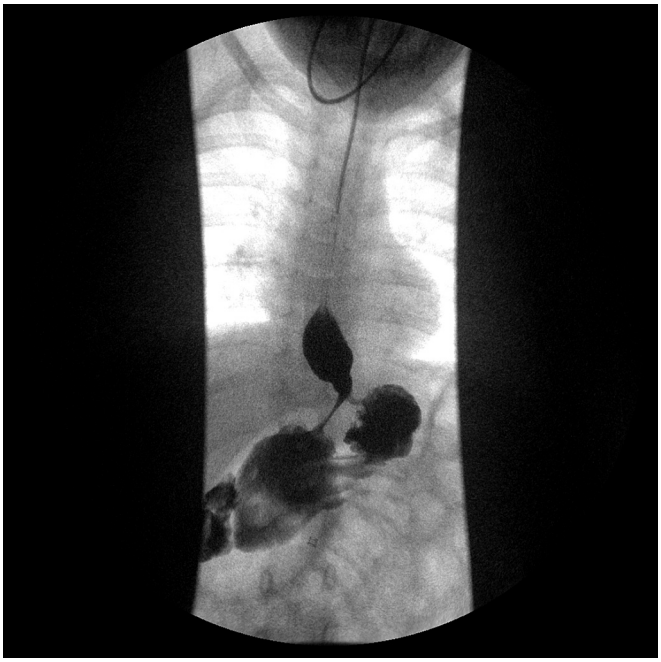

Fig. 1. Contrast esophagogram demonstrating Y-shaped deformity of the distal esophagus.

computed tomography scan ruled out tumors, abscesses, and foreign bodies.

1.3. Surgical intervention

She then underwent a repeat open Nissen fundoplication. During the operation the esophagus was found to bifurcate into two independent outlets near the gastroesophageal junction. The medial outlet was identified as the physiologic esophagus while the smaller diagonal outlet was identified as the fistula connecting the distal esophagus with the gastric fundus (Fig. 2). It was also noted that the previous fundoplication had partially unwrapped. During the repeat Nissen Fundoplication the fistula between the distal esophagus and gastric fundus were separated. The esophageal and the gastric aspects of the fistula were repaired using two layers of 3-0 silk suture. A 32 French bougie was placed in the esophagus and a posterior crural repair was done with 3 interrupted 2-0 silk sutures. The greater curvature of the stomach was then wrapped 360° around the native esophagus and secured with four additional 2-0 silk sutures to maintain tissue control.

1.4. Postoperative

Postoperatively, she had dysphagia and chest pain. Repeat contrast swallow showed moderately tight fundoplication requiring two esophageal bouginages. On follow-up the patient's symptoms had resolved, she is now thriving.

2. Discussion

Esophagogastric fistulas are rare and have been described in association with peptic inflammation in the distal esophagus [1,2], Crohn's disease [3], neoplasia [4], and surgical procedures of the gastro-esophageal junction [4–7]. This is particularly true in patients with repeated operations with history of scleroderma [5,6]. Choudhry and colleagues were the first to distinguish between two forms of the disease [6]. Esophagogastric fistula is described as a

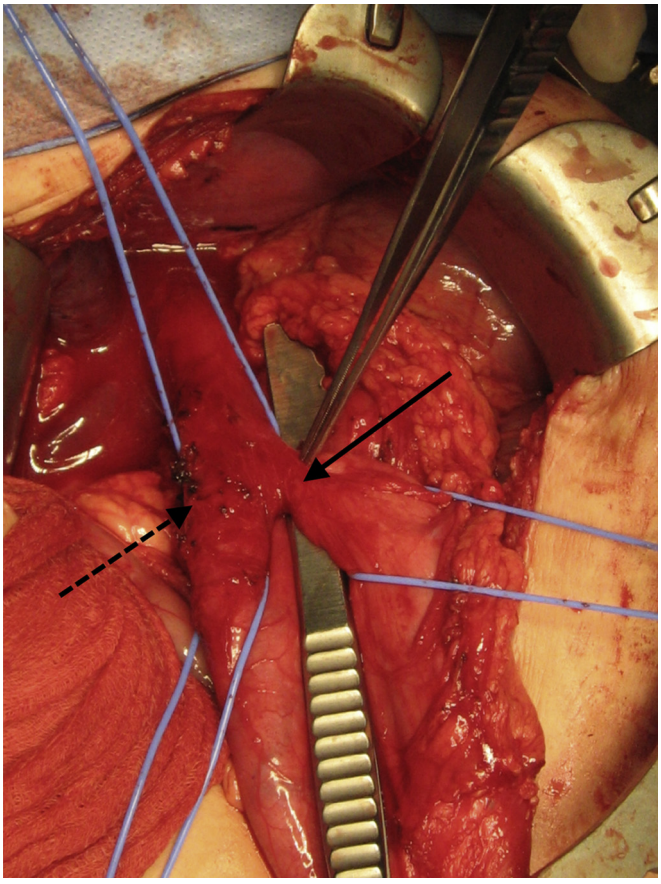

Fig. 2. Intraoperative image demonstrating native esophagus (dashed arrow) versus gastroesophageal fistula (line arrow).

narrow fistula between the distal esophagus and gastric fundus; whereas a "double-lumen esophagus" refers to a fistula of equal or similar size to the physiologic esophagus, as in our case [6].

Several mechanisms of esophagogastric fistula formation have been postulated. The most prominent one being the persistence of gastro-esophageal reflux induced ulcers, which erode the gastric fundus allowing it to anastomose with the distal esophagus [4–6]. Another possible mechanism involves surgically induced ischemia caused by strangulating sutures resulting in tissue necrosis [5,6]. A third possible mechanism is a full thickness suture "bite" causing a perforation which may develop into a fistula. There is also one reported case of esophagogastric fistula being caused by pledgets eroding through the esophagus and gastric fundus after laparoscopic Nissen fundoplication [7]. The primary mechanism responsible for the formation of esophagogastric fistulas is still unclear due to the rare nature of the disease.

Prior to surgical intervention our patient had signs of gastro-esophageal reflux visible on upper endoscopy. This most likely was caused by full thickness suture "bite" perforation during the first fundoplication which resulted in a fistula. We postulate that severe reflux and adenotonsillar hypertrophy occurred as gastric contents traveling thru the fistula and into the esophagus causing odynophagia and nocturnal cough. This hypothesis is supported by the severe erosive esophagitis identified near the fistula.

Conservative management with dilation and proton pump inhibitors was not successful in this patient. Failure of conservative management made her a candidate for surgical intervention. After fistula repair and repeat Nissen fundoplication was preformed, she complained of dysphagia and chest pain. We believe these

symptoms were related to postoperative edema, tightness of the new fundoplication, and possible dysmotility secondary to surgical dissection and removal of fistula. A contrast study postoperatively indicated a moderately tight fundoplication with gastric and esophageal integrity. Two esophageal bougienages were preformed which ultimately resolved her postoperative dysphagia.

The true incidence of esophagogastric fistula after fundoplication is unknown and it is unclear whether every case requires formal repair. In the case described, the fistula was present on the first contrast study after the original fundoplication when the patient was 2 years old. Repeat surgical intervention was initiated four years later as the patient's symptoms worsened.

There is no consensus regarding the best treatment for this rare condition. In a case report of esophagogastric fistula caused by Crohn's disease, the fistula resolved after treatment of underlying disease [3]. In two other cases acid suppression therapy with proton pump inhibitors was successful in treating the symptoms, however, the fistulas persisted [1,8]. Of the other cases in the literature, there is no documentation of resolution of esophagogastric fistula without surgery. In particular, no post-Nissen fistula resolved without surgical repair. Raymond et al. described treatment of a peptic fistula with antacids, but provided no information about long-term outcomes [2]. Choudhry and colleagues [6] reported two cases, one post complex repeat fundoplication and one with no prior surgery. Both were managed non-surgically with esophageal bougienage; neither patient had a resolution of the fistula [6]. Fleming and DiMagno [4] described a fistula which occurred after complex repeat gastric surgery and Nissen fundoplication. The patient had conservative management without resolution of the fistula [4]. Mullen et al. [5] reported two cases which occurred after Nissen fundoplication, one was re-operated and resolved. The other was not repaired and persisted [5]. Hussain et al. [9] described a case of a 17 year old female with a history significant for laparoscopic Nissen Fundoplication at the age of 2 then an open revision 3 years later. Conservative management was unsuccessful; however, surgical management resolved her symptoms, even improving her appetite and increasing her weight by 20%.

3. Conclusion

Esophagogastric fistulas although rare may arise as a complication to Nissen fundoplication. Surgical intervention is the only definitive treatment in patients with iatrogenic esophagogastric fistulas.

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